



Kasık Ağrısı ve Topallama ile Ortaya Çıkan Pelvisin Primer Kist Hidatiği: Olg u Sunumu

Primary Cyst Hydatidosis of Pelvis with Groin Pain and Limb: a Case Report

Kasık Kist Hidatik / Pelvis Cyst Hydatidosis

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Özet

Ekinokok vücutta tüm organları tutabilir. Ancak kasıkla birlikte kemiği tutan ve anterior kalça ağrısına neden olan primer pelvik kist hidatik çok nadirdir. Bu olguda kemiği tutmuş ancak patolojik kırığa neden olmamış yaygın pelvik kist hidatiği olan 22 yaşındaki erkek hastayı sunmaktayız. Hastanın başlangıç şikayeti kasık, kalça ve bacak ağrısı idi. Hastanın tanısı klinik ve radyolojik olarak konuldu ve hasta en blok rezeksiyon ve kemik sementleme ile tedavi edildi. Hasta cerrahiden bir yıl sonra tamamen iyileşti. Pelvik kist hidatik üriner, genital ve abdominal sorunlara yol açmasına rağmen kasık ve kalça anteriorunda ağrısı bulunan hastaların ayırıcı tanısı içinde bulunmalıdır. Çoğu kist hidatik antihelmintik ilaçlarla tedavi edilebilmektedir ancak kemiğin etkilendiği olgularda rekürrens ve kemik kırığından kaçınmak için küretaj ve kemik sementleme iyi bir seçenektir.

Anahtar Kelimeler

Kist Hidatik, Pelvis, kemik, Kasık Ağrısı.

Abstract

Echinococcosis may involve all organs of body; however, primary pelvic cyst hydatid involving bone with groin and anterior hip pain is very rare. We presented a case of extensive pelvic cyst hydatidosis involving bone without pathological fracture, in a 22-years-old man. First symptoms of patient was groin and anterior hip pain and limb. The diagnosis was made clinico-radiologically and the patient was cured with enbloc resection and filling bone cement. The patient was relieved completely one year after surgery. Cyst hydatid should be included in the differential diagnosis in patients with groin and anterior hip pain, although pelvic cyst hydatoid result in urinary, genital and abdomen problems. Much hydatid disease can be cure by administration of antihelmintic drugs, however, curettage and bone cementing is good choice in patient involving bone to eliminate recurrences and bone fracture.

Keywords

Cyst Hydatid, Pelvis, Bone, Hip Pain.

Introduction

Cyst hydatidosis is a parasitic infection disease caused by larval stage of *Echinococcus granulosus* or less commonly *Echinococcus multilocularis*. *Echinococcus granulosus* is the most common form of the cyst hydatidosis and is endemic in the South America, Mediterranean, and Middle East. Cysts of Echinococcal are mostly existed in the liver (60%-70%), followed by the lungs (10%-25%), spleen, ovaries, kidneys, brain, bones and heart[1]. Echinococcosis may involve all organs of body; however, primary pelvic cyst is very rare [2,3]. Hydatoid disease in extrahepatic locations usually remains asymptomatic unless the cyst grows and causes symptoms due to pressure, rupture to peritoneal cavity, secondary infection, an allergic reaction or irritation of periost[1]. Pelvic cyst results in to urinary, genital, and abdomen problems usually[2]. It may come out with hip pain rarely[4].

Osseous echinococcosis is very rare and occurs in only 0.5-4% of all the cases of Cyst Hydatidosis. Spine and pelvis are most common bones attached by echinococcosis. In addition to, it may involve the femur, tibia, humerus, skull, and ribs [4]. The initial location of the lesion in long bones is metaphyseal or epiphyseal, later extending to the diaphysis by hematogenes. It remains asymptomatic and result in pathologic bone fractures. Prognosis of bone echinococcosis is poor and it does not respond to the medical treatment. Surgical excision is required usually.

We present a 22-years-old male patient with groin and anterior hip pain due to pelvic cyst hydatid involving acetabulum and pubic ramus superior without fracture. Pelvic cyst hydatoid similar to our case is very rare in the literature.

Case

A 22-years-old man presented with right groin and anterior hip pain for 4 months duration. He had also suffered from limping due to painful leg during this period. Plain X-Ray of the pelvis showed destructive lesion in acetabulum and the superior pubic ramus (Figure 1). Routine blood count was normal. There was a cystic lesion destroying the cortex of acetabulum on CT (Figure 2). MRI showed multiple cystic lesions in the superior and inferior pubic ramus and retrorectal space, but lesion did not involve the hip joint (Figure 3). Chest tomography and ultrasound of liver was normal. In surgery, a hydatid cyst was found in the pelvis involving the acetabulum and spreading retrorectal space. Cyst was taken out as an en bloc. Curettage swabbing with hypertonic saline were carried out and the defect was filled with polymethylmetacrylate (Figure 4). After the operation, pathology report that the cyst was hydatid cyst. Patient was treated with oral Albendazole for 3 months with a dose of 60 mg/kg/day after surgery. He has been well one year after surgery.

Discussion

Echinococcus granulosus is a 5-mm long worm, with a lifespan of 5-20 month in the intestinal system of wild animals. When eggs

come to human intestinal system, they enter the portal circulation, and are then catch in the liver. If they run away from the hepatic filter system, they join the systemic circulation and seat in the liver, lungs, kidney or in other organs.

The incidence in men was slightly less than twice that in women, and the mean age of presentation of the patients was 51 years. No cases were recorded in children under the age of 8 years. As hydatid disease of bone remains asymptomatic over a long period, It can be diagnosed late decades. Our patient was male and age was 22. This age was earlier than other cases in literature.

Pelvic involvement is usually secondary to hepatic or pulmonary hydatidosis, however it may on occasion occur as the primary disease[5]. Ladeb et al reported that only 2% of cystic hydatid disease cases were localized in the pelvis [6]. Similarly, the incidence of primary pelvic cyst hydatid in Turkey is rare (2%) [7]. Our case was also isolated hydatid cyst of the pelvis involving bone.

The symptoms of pelvic echinococcosis are not specific and may involve abdominal pain, swelling, menstrual irregularities, infertility, obstruction of labor and pressure symptoms from adjacent organs including the bladder and rectum. Belzunegui J et al reported case of cyst hydatid with hip pain involving pelvis, hip joint and femur. In our patient, the primary complaint was groin and anterior hip pain and limb [8]. They treated hydatidosis with surgery "Girdlestone arthroplasty". Morris BS et al presented a patient with pelvic cyst hydatid suffering hip pain. Our patient's first symptom was groin and anterior hip pain similar to last two cases [4].

USG is the first option tool initial diagnostic, but CT is more efficient showing calcification and daughter cysts, and is therefore more sensitive than US for the differential diagnosis of cyst hydatoid. MRI shows both soft tissue and bone components. A definite preoperative diagnosis without histological examination is often difficult as there are no pathognomonic signs, radiological findings may be confused with those of other tumoral lesions, and serological tests are of limited value. Difficulty in both diagnosis and management are hallmarks of hydatidosis of bone. The disease mimics chronic osteomyelitis, fibrous dysplasia of bone osteosarcoma, benign cystic lesion of bone, brown tumor (hyperparathyroidism), and various other neoplastic lesions. Lesion in our patient has both soft tissue and bone component. We used to X-Ray, CT and MRI to make a diagnosis. It was difficult to make a differential diagnosis[1,4].

Osteohydatid disease has been considered to change between 1 and 4%. Bone cyst hydatoid involves the spine in 35% of cases, the pelvis in 21% of cases, the femur in 16% of cases and the tibia in 10% of cases in literature. The ribs, skull, scapula, humerus and fibula harbour cysts with an incidence in decreasing order of frequency (6 and 2% of the total cases of bone hydatid). Hydatid



Figure 1. Cyst hydatoid on x-ray.

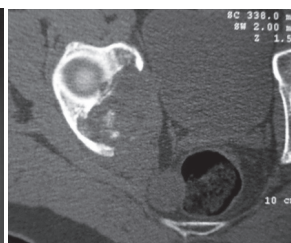


Figure 2. Cyst destroyed the bone cortex.

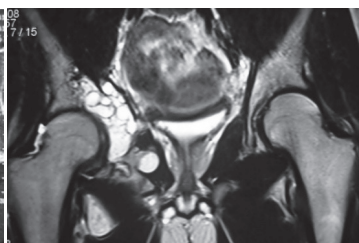


Figure 3. Bone and soft tissue component of cyst hydatoid.

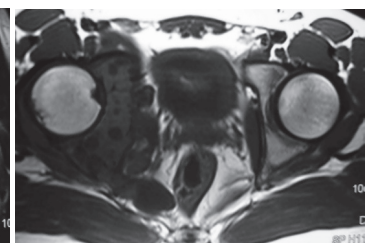


Figure 4. After en bloc resection of cyst and filling bone cement on x-ray.

cysts of bone may remain asymptomatic over a long period, however, echinococcus may destruct bone and may result in severe pain and bone fracture. When a pathological fracture occurs in long bones due to hydatid cyst, non-union is common. Acetabulum was destroyed by echinococcus in our patients. Pathologic fracture did not occur due to early diagnosis.

Treatment for extrahepatic echinococcal disease is based on size, location and manifestations of the cysts, and the overall health condition of the patient. Asymptomatic small cysts once diagnosed can be treated with antihelminthic drugs, administered for 6 months. For symptomatic or large hydatid peritoneal cysts, surgery is the first option of treatment. Surgical treatment can be either radical or conservative. Total cystectomy is the best method. It should take care that the abdominal and pelvic cavity is isolated with gauzes soaked in 20% hypertonic saline solution to avoid secondary hydatidosis and allergic reaction [5]. Preoperative 3 months albendazole administration decreases the viability of the hydatid cysts and may reduce the risk of anaphylaxis, but the

time of the treatment is not clear [4,6]. In addition it should be continued for a month after surgery. We have no opportunity to use preoperative, but we used albendazole for 3 months postoperative. Osseous hydatidosis should be treated with radical resection with a wide margin of healthy tissue. This may be difficult, but incomplete removal results in recurrence. We did radical resection of soft tissue lesion and curet the bone lesion. Then bone defect was filled with polymethylmetacrylate. Although a review of the literature reveals a poor prognosis if the disease is extensive in the pelvis, our patient had no recurrence at one year follow up.

In conclusion, cyst hydatid should be included in the differential diagnosis in patients with groin and anterior hip pain, although pelvic cyst hydatoid result in urinary, genital and abdomen problems. Much hydatid disease can be cure by administration of antihelminthic drugs, however, curettage and bone cementing is good choice in patient involving bone to eliminate recurrences and bone fracture[8].

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